Embolization of Vascular Malformations in Head and Neck Regions

A Single Center Experience

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Summary

The purpose of this study is to determine the effectiveness of embolization of each type of vascular malformation.

Thirty three patients with a diagnosis of vascular malformations in head and neck regions who were treated by embolization at Siriraj Hospital, Thailand, between 1997-2002 were reviewed retrospectively. There were 19 arteriovenous malformations (AVMs), two arteriolar-capillary types, ten venous malformations, one veno-lymphatic malformation and one mixed capillary and venous types. The goal of treatment is to restore function and to prevent bleeding complications with particularly good cosmetic results. The technique and therapeutic agents depended on the types and flow characteristics of each malformation. Transarterial embolization with N-butyl cyanoacrylate were used in AVMs, unless no arterial route approach, then direct puncture was tried. This was carried out in five patients with AVMs. Polyvinyl alcohol was used in two capillary lesions. All venous malformations were treated by absolute alcohol injection, percutaneously. All had good outcome without serious complications. Two patients had further plastic surgery, one was AVM of eyelid and the other was facial venolymphatic malformation. Embolization is considered to be the primary treatment by eradication of those abnormal vessels directly at the target, whereas delayed plastic surgery may be needed later.

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Introduction

The most accepted classification of congenital vascular anomalies was described by Mulliken and Glowacki in 1982 [1]. This classification differentiates haemangiomas from vascular malformations on the basis of endothelial cell characteristics and the clinical behavior. Haemangiomas are the most common tumors of infancy, predominantly in females. They tend to be small or absent at birth but show increased mitotic activity during the proliferating phase shortly after birth. They then undergo regression in the involuting phase, which is usually complete by age 5-7 years. Conversely, vascular malformations are usually noted at birth, do not regress but rather grow commensurately with the child. They are structural abnormalities, error of vascular morphogenesis, and display normal endothelial cell mitotic activity.

Enlargement can occur with changes in pressure and flow, ectasia, collateral formation, shunting and hormonal modulation. Vascular malformations may have any combination of capillary, venous, arterial and lymphatic components with or without fistula [1,2].

The classification scheme for vascular anomalies was modified by the International Society

for the Study of Vascular Anomalies (ISSVA) in 1992[3]. The vascular malformations are classified into simple forms which are included capillary, venous, lymphatic and arteriovenous malformations (AVM) and complex-combined forms. They may be slow-flow or high flow lesions. The choice of therapy depends on type, site and extent of the malformation. The purpose of this study is to determine the effectiveness of embolizations for treatment of vascular malformations. The approach and therapeutic agents were depended on the types and flow characteristics of each malformation.

Table 1 **Anatomic locations of vascular malformations**

Sites	AVM	Capillary	Venous
Ear	4	-	1
Ear and neck	2	-	-
Nose	1	-	-
Lip	2	-	2*
Tongue	-	-	3**
Face	3	1	6***
Mandible	3	-	-
Scalp	2	-	-
Pre-auricle	-	1	-
Eyelid	2	-	-
Total	19	2	12

** One patient had both lip and tongue lesions.*
*** One patient had combined capillary and venous malformation.*
**** One patient had combined venous and lymphatic malformation.*

Material and Methods

A retrospective review was performed between January 1997 and December 2002 on 33 patients (15 male and 18 female) ranging in age from nine to 51 years (mean age = 28 years) with diagnosis of vascular malformations in head and neck regions who were referred to our radiology department for embolization. Diagnoses were based on clinical presentation and physical examination.

The presenting signs and symptoms, in order of frequency, included cosmetic problems, distortion of adjacent structures, pain, functional limitation, especially the lesions of lip and tongue and bleeding. Pretreatment evaluation was individualized for each patient. The computed tomography and/or magnetic resonance imaging were performed in some cases when needed, particularly in patients with venous malformations to determine the type and boundaries of lesions and clarify appropriate sites for percutaneous puncture. Diagnostic arteriography was not indicated if the clinical diagnosis of venous malformation was obvious. The goal of treatment is to restore function and to prevent bleeding complications with particularly good cosmetic result.

The procedures were performed under general anesthesia. The treatment of choice for AVM was transarterial embolization using N-butyl cyanoacrylate (NBCA), if no arterial route could be accessed, then direct puncture was tried. Transarterial embolization by polyvinyl alcohol (PVA) was used in case of capillary malformation. Percutaneous injection of

Table 2 **Types of treatments**

Treatments	AVM	Capillary	Venous	Capillary-venous	Venous-lymphatic
Transarterial NBCA	14				
Percutaneous NBCA	1				
Transarterial plus Percutaneous NBCA	4				
Transarterial PVA		2			
Percutaneous Alcohol			10		1
Transarterial PVA plus Percutaneous Alcohol				1	
Total	19	2	10	1	1

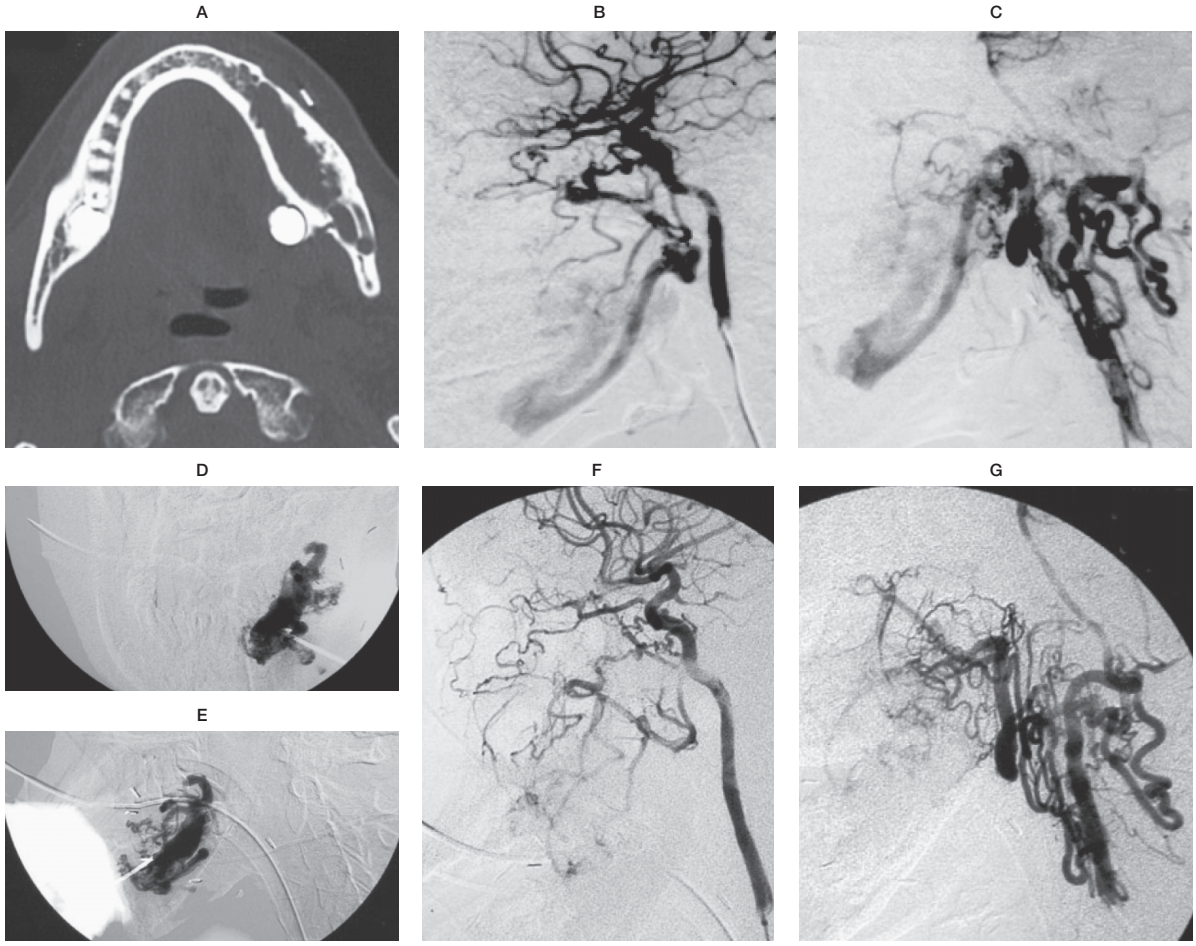

Figure 1 Intraosseous AVM of mandible in a 16-year-old female who presented with massive bleeding per gum status post external carotid artery ligation. A) Axial CT scan revealing osteolytic appearance of the malformation. B) Left internal carotid angiogram exhibiting reconstitution of the internal maxillary artery from anastomosis of ILT with consequence supply to the AVM via inferior alveolar artery. C) Recruitment of the external carotid system from anastomosis of vertebral artery too. D) Frontal and E) Lateral projection during NBCA embolization via direct transosseous puncture. F) Left internal carotid angiography and G) Left vertebral angiography immediate post embolization demonstrating complete obliteration of the malformation.

sclerosing agent was performed in all venous malformations.

The lesions were directly punctured by means of palpation or imaging guidance. Mixture of absolute ethanol and nonionic contrast medium (1:2 by volume) was used to render radiopaque sclerosing agent in all cases. The injection was performed under fluoroscope, after test injection by contrast media in each venous space. Manual compression of venous outlet was placed when possible, to minimize passage of alcohol into systemic circulation and to maximize the sclerosing time.

The end point of injection was when visualization of the venous outlet was recognized. A maximum volume of ethanol in each one sitting was 1 ml/kg body weight which was not exceeded 40 ml. In most lesions, multiple access sites were required. Repeated embolization for all patients were planned after 4-6 weeks follow-up, if needed, it was recommended approximatedly in next 12 weeks. Surgery was considered individualized for each patient, if possible, after finished embolization and the result was not so satisfactory.

Results were considered to be cure when the lesions became clinically undetectable after more than 12 weeks of treatment, and to be satisfactory when the sizes were reduced at least by 50% or no events of the complications of the diseases such as bleeding, pain, after more than 12 weeks of treatment.

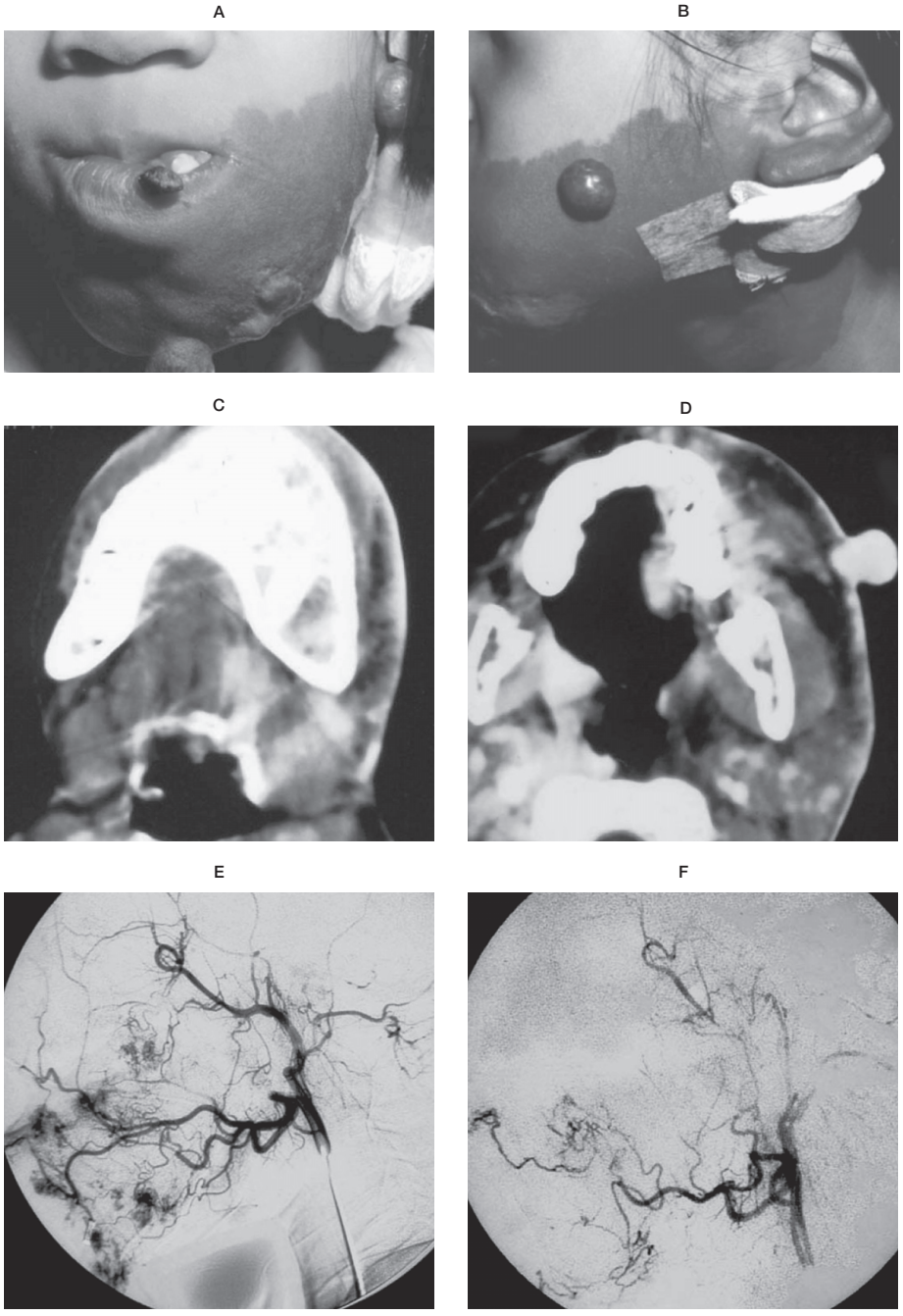

Figure 2 Capillary malformation in lower face of a 7-year-old girl presenting with bleeding complication.
A,B) Photographs showing redness and nodular ectasias of the skin with gingival hypertrophy.
C,D) Axial CT scans demonstrating hypertrophy of the mandible and underlying soft tissue.
E) Left external carotid angiography revealing multiple nodular capillary stainings.
F) Post transarterial embolization with polyvinyl alcohol exhibiting obliteration of those capillary stainings.

Results

There were 19 AVMs (57.6%), two capillary malformations (6%), ten venous malformations (30.3%), 1 veno-lymphatic malformation (3%) and one combined capillary and venous types (3%). The anatomic locations of each type of lesions were shown in table 1.

The type of approach or access and embolic agents were based on the diagnosis according to the classification as demonstrated in table 2. For the AVMs, transarterial embolizations using NBCA were approached in 14 patients, combined transarterial approach and percutaneous puncture directly into the lesions were performed in four patients.

One patient who had intraosseous AVM of the mandible and had previous surgical ligation

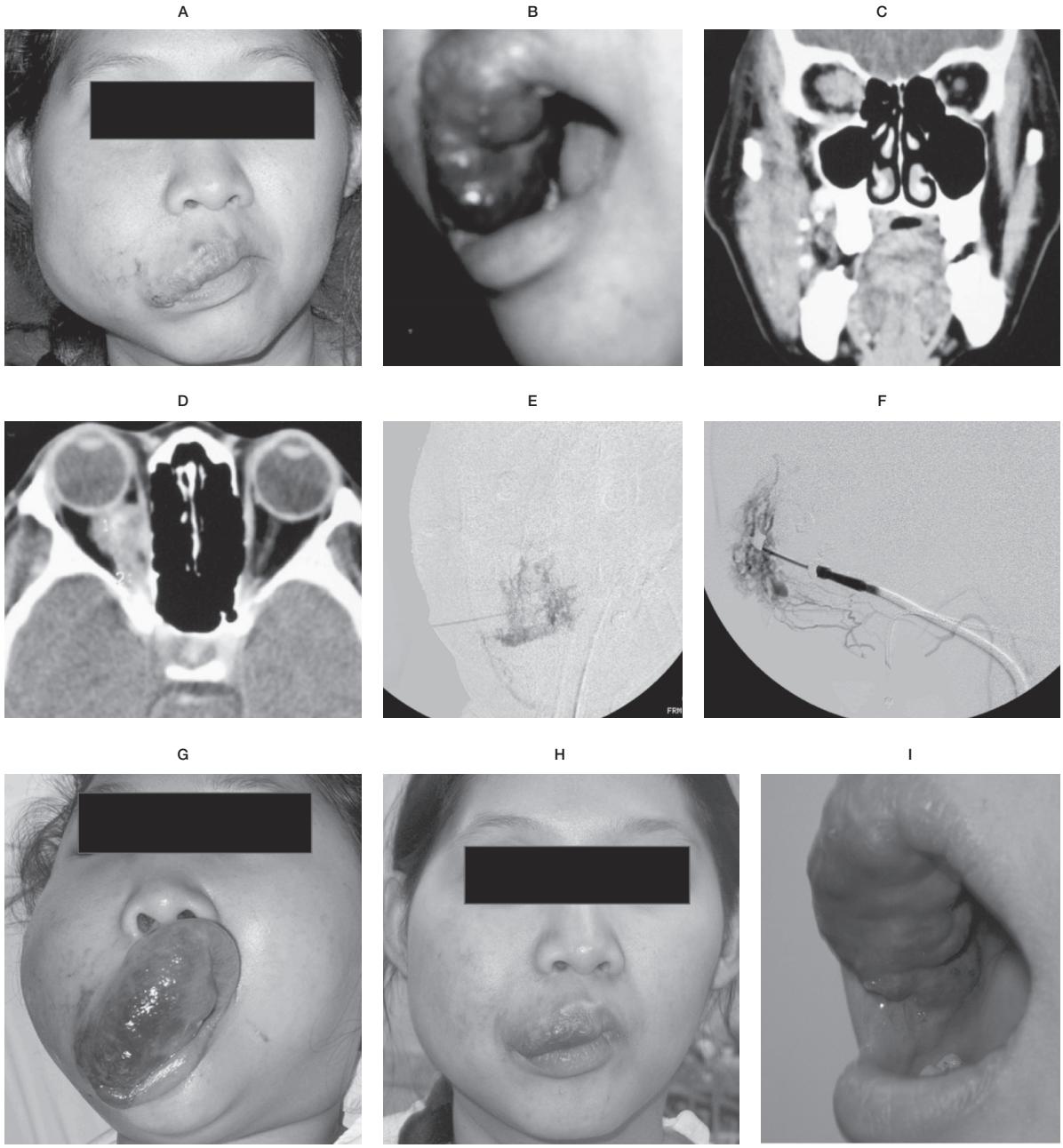

Figure 3 A 16-year-old female with classical presentation of venous malformation. (A,B) Venous malformation involving right check, buccal area and upper lip. C) Noncontrast coronal and D) Noncontrast axial CT scans showing extension of the lesion with involvement of right orbit. Note multiple phelboliths inside the lesion. E) Frontal and F) lateral projections during direct puncture with injection of mixture of ethanol and contrast media. G) Photograph taken one day after sclerosing therapy. H,I) Significant reduction of venous malformation after four sessions of injections (compare to A and B).

of the external carotid artery was directly punctured into the lesion of the bone by using CT imaging guidance and embolized by NBCA (figure 1).

Only two patients had capillary malformations which exhibit capillary blush on angiogram. One had the lesion in lower face with mandibular and gingival hypertrophy. Palliative embolization with polyvinyl alcohol was performed (figure 2). Unfortunately, the patient lost to follow up before reconstructive surgery was planned. The other patient had small lesion

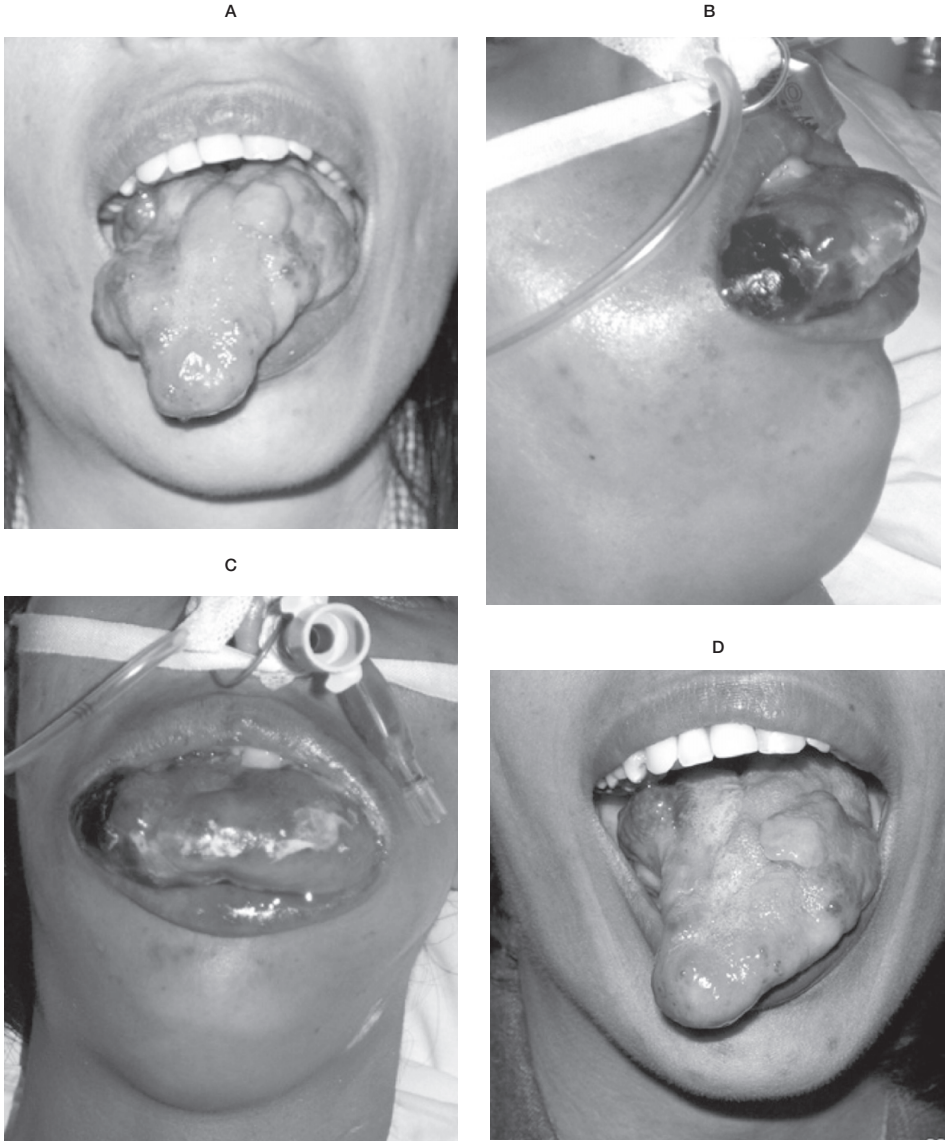

Figure 4 Venous malformation involving tongue and floor of mouth.
A) Pre-treatment.
B,C) 2 days after ethanol injection of the lesion at right side of tongue and at floor of mouth. Note severe swelling of the tongue and necrosis of the lesion. The patient needed prolonged intubation and prophylactic tracheostomy.
E) 3 months after sclerotherapy.

of palpable mass at preauricular area with complete obliteration after embolization.

Ethanol was used as sclerosing therapy for all venous malformations by means of percutaneous punctures. Most cases had multiple punctures in multiple sessions, depending on their size. The limitation was the volume of alcohol at its maximal amount for each time. Immediate swelling were exhibit in all patents with maximum at day 2 or 3, then subsequently resolution in about two weeks (figure 3). In one patient who had venous malformation of tongue, prolonged intubation with prophylactic tracheostomy was decided to protect the airway, because of severe swelling of the tongue (figure 4).

Table 3 **Outcome of treatment**

Outcome	No (%)
Cure*	12 (36.4%)
satisfactory	21 (63.6%)
Complications	3 (9%)
Total	33 (100%)
** Post embo surgery 2*	

In all venous malformations, no skin necrosis or ulceration were found. One patient who was an 8-year-old boy presenting with bulky red mass at base of tongue with compromised pharyngeal airway, was classified to be mixed capil-

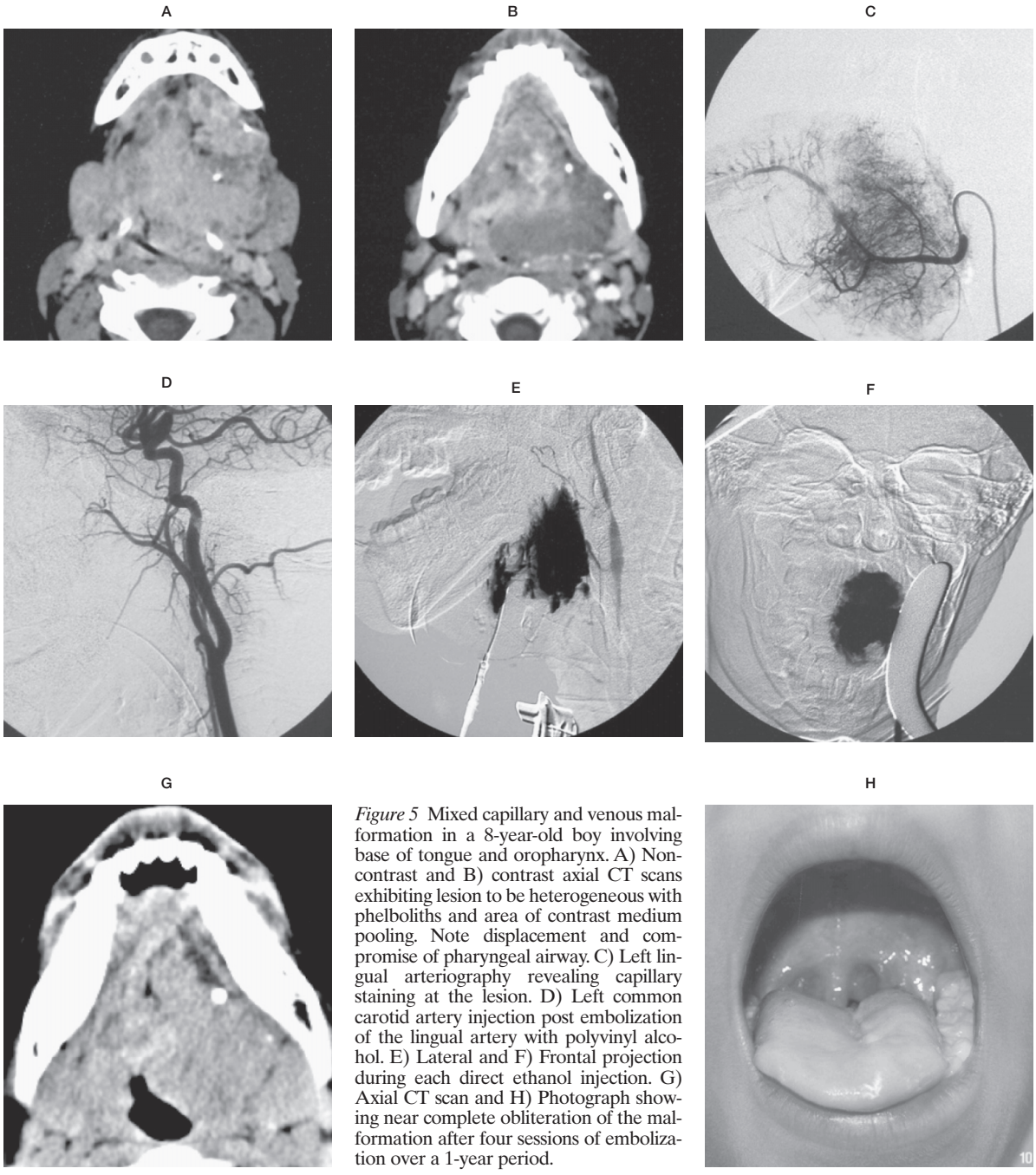

Figure 5 Mixed capillary and venous malformation in a 8-year-old boy involving base of tongue and oropharynx. A) Non-contrast and B) contrast axial CT scans exhibiting lesion to be heterogeneous with phelboliths and area of contrast medium pooling. Note displacement and compromise of pharyngeal airway. C) Left lingual arteriography revealing capillary staining at the lesion. D) Left common carotid artery injection post embolization of the lingual artery with polyvinyl alcohol. E) Lateral and F) Frontal projection during each direct ethanol injection. G) Axial CT scan and H) Photograph showing near complete obliteration of the malformation after four sessions of embolization over a 1-year period.

lary and venous type by CT imaging showing multiple venous spaces with phelboliths at base of tongue and submandibular space whereas the angiogram showed capillary stain from left lingual artery. After the airway was secured, transarterial embolization of the lingual artery by using polyvinyl alcohol was tried first then followed by multiple sessions of percutneous injection of ethanol in the submandibular spaces and directly in the residual mass at base of tongue (figure 5). The outcome was considered to be cure.

Of total 33 patients, cure was documented in 12 patients (36.4%) and satisfactory in 21 patients (63.6%) (table 3). Two in 12 patients who were cured had post embolization reconstructive surgery, one was combined type of veno-lymphatic malformation of face and the other

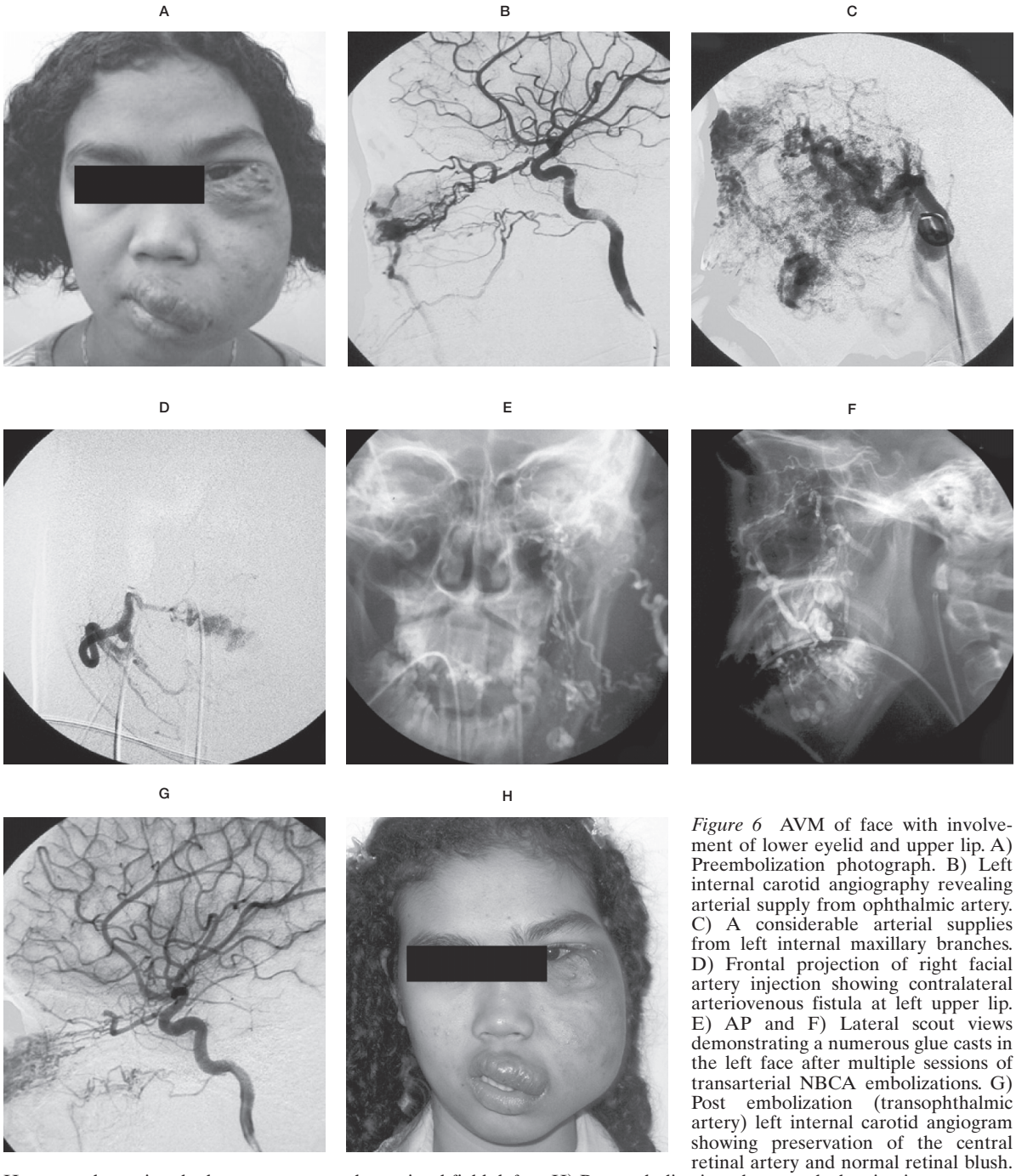

Figure 6 AVM of face with involvement of lower eyelid and upper lip. A) Preembolization photograph. B) Left internal carotid angiography revealing arterial supply from ophthalmic artery. C) A considerable arterial supplies from left internal maxillary branches. D) Frontal projection of right facial artery injection showing contralateral arteriovenous fistula at left upper lip. E) AP and F) Lateral scout views demonstrating a numerous glue casts in the left face after multiple sessions of transarterial NBCA embolizations. G) Post embolization (transophthalmic artery) left internal carotid angiogram showing preservation of the central retinal artery and normal retinal blush. However, the patient had upper outer quadrant visual field defect. H) Post embolization photograph showing improvement of the malformation, particularly that of upper lip.

patient had AVM of eyelid. Three patients (9%) had complications. One patient who had AVM of hemiface had quadrant visual field defect 24 hours after embolized through the ophthalmic artery, although the tip of the microcatheter positioned beyond the origin of central retinal artery, and immediate controlled angiogram showed patency of the central retinal artery as well as normal retinal blush (figure 6). One patient had facial palsy after percutaneous injection of ethanol in venous malformation of cheek.

The other patient had transient locked jaw with ethanol injection of the lesion of tongue and submandibular area.

Discussion

Proper management of vascular anomalies must begin with precise terminology and classification [4]. Vascular malformations demonstrated no tritiated thymidine incorporation and normal ultrastructural characteristics. They consisted of abnormal, often combined, capillary, arterial, venous and lymphatic vascular elements [1].

Arteriovenous malformations in the head and neck region are far less common than intracranial AVMs [4]. Fast- flow typically become evident in childhood. Puberty or trauma seem to trigger expansion [4,5]. Treatment of these lesions is prompted by clinical symptoms that may vary with their type, size and locations. They usually are cosmetic problem, pain, bleeding or ulceration. Selective angiography is better to characterize nidus, flow pattern, micro or macroarteriovenous fistula, preceeding interventional therapy.

Occlusion of the nidus and/or fistula by transarterial catherization has been used successfully in the management of AVMs as the sole therapy [4-6]. Although there have been many reports of a wide variety of embolic agents for transarterial embolization [7-10], we have confidence to use N-butyl cyanocrylate (NBCA) because of its proved property and effectiveness [5,11].

When there is a limited role for transcatheter arterial embolization in patients with previous proximal arterial ligation or small arterial supplies but large venous pouch, percutaneous puncture is an alternative method [6,12]. We used CT imaging and road mapping angiography as the guided punctures. We have performed in five of 14 AVMs. Two of them were mandibular AVMs which were cured, one was huge AVM of ear and neck region post transarterial embolization and reconstructive surgery but inadequate. No complications was found by this approach. As the primary treatment of AVMs, ligation or proximal embolization of the feeding arteries should never be performed. Rapid recruitment of flow from normal anastomosis of the head and neck region will then supply the nidus, furthermore, proximal arterial blockage denies access for embolization [4,13].

Capillary malformations are composed of dilated capillary-to-venular-sized vessels in the superficial dermis. With age, the vessels gradually dilate with tendency to nodular ectasia, often associated with hypertrophy of soft tissue and underlying bone [4]. Embolization is indicated for palliation when complication such as bleeding is presented or for preoperative correction of soft tissue and skeletal hypertrophy. It has been noted that a capillary malformation can be a red flag signaling an underlying structural abnormality [4], but this was not evident in our two cases of capillary malformations.

Venous malformations are most common vascular malformations, present at birth, but they are not always evident. They grow proportionately to the child, expand slowly and often enlarge during puberty. They are bluish, soft, compressible and expand when the affected area is dependent or after a Valsalva maneuver. Phelboliths are commonly found [4,5,14,15]. Direct phlebography is performed to comfirm diagnosis and exclude other soft-tissue tumor, particularly, it plays an important role as part of sclerotherapy. Absolute alcohol is the most commonly used agent with proved to be an excellent sclerosing agent that causes complete vascular occlusion. Ethanol denatures blood proteins and injures vascular endothelium, inciting an intense thrombosis.

In addition, it is readily available, inexpensive and easy to administer [15-17]. Alcoholic solution of zein (Ethibloc) sodium morrhuate, ethanolamine and sotradecol have been reported as effective sclerosing agents [15,18]. In our experience, we have used only absolute alcohol in treatment of venous malformations. Although it was mixed with nonionic liquid contrast medium at 2:1 by volume, without precipitation, it has still provided good effectiveness and good result. Furthermore, the advantage is visualization of the alcohol injection during fluoroscopic control.

The main complications of sclerotherapy are cutaneous necrosis and neural toxicity. Systemic complications area rare and are related to the passage of alcohol into the systemic circulation. These include hemolysis with potential renal toxicity and cardiac arrest. So it is important to be certain that no extravasation during injec-

tion. A manual compression of venous outlet or jugular vein is useful to minimize passage of alcohol whereas increase sclerosing time.

The alcohol induces a significant inflammatory reaction that may worsen the symptoms, then analgesics and anti-inflammatory agents should be given. Particularly in patients with lesions that may cause compromised airway, such as base of tongue, oropharynx or laryngeal area, prolonged intubation or tracheostomy with intensive care must be considered.

Conclusions

A plan of treatment of vascular malformations based on their flow characteristics. The precise terminology and classification will clarify the dynamics within the lesion. A better understanding of the pathogenesis and natural history will provide corrected diagnosis and well-reasoned treatment plan. These patients deserve to be cared for by a multidisplinary team in a common language.

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